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IMAGING IN THORACIC CANCER

Esophageal cancer complicated by esophagopulmonary fistula and lung abscess formation: A surgical approach

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Introduction

Esophageal cancer is an aggressive malignancy with an unfavorable prognosis and an increasing incidence worldwide.[1] Locally advanced esophageal carcinoma might eventually be complicated by esophagorespiratory fistula, which is predominantly palliated by endoscopic stent placement.

A subgroup of these patients present with direct tumor invasion of the adjacent pulmonary tissue, resulting in esophagopulmonary fistula and subsequent pulmonary abscess formation.[2] Despite stent placement, the prognosis of esophagopulmonary fistula is grim. Most of these patients suffer from persistent pulmonary abscess formation leading to deterioration in their clinical condition and a fatal outcome within a short time.[2,3] Therefore, the therapy of patients with esophagopulmonary fistulas should be focused on pulmonary abscess formation, which requires a different approach than that used for patients with uncomplicated esophagorespiratory fistulas.

Surgical treatment with resection of the esophageal lesion and the involved pulmonary lobe might be a more promising option than conventional stent placement because this strategy enables the removal of all inflammatory tissue and might significantly prolong the survival time of these patients.[4–6]

Despite the potential benefits, it seems that most surgeons are rather reluctant to perform major surgery in these patients, who generally present in poor health.[7] To our knowledge, there are no current reports addressing the issue of surgery for esophagopulmonary fistulas, apparently reflecting the widespread nihilistic attitude towards this life-threatening condition.

Not only is the incidence of esophageal cancer constantly rising, but neoadjuvant radio-chemotherapy protocols are also being used more frequently, and these have been previously associated with the formation of esophageal fistulas.[8] Consequently, the number of patients presenting with intractable esophagopulmonary fistulas is likely to increase, challenging the treatment modalities available.

In our view, this problem is not adequately reflected in the current literature. Therefore, we report our experience with the surgical treatment of two consecutive patients presenting with malignant esophagopulmonary fistulas, who underwent simultaneous esophageal and pulmonary resection.

Patients

A 65-year-old severely cachectic woman (body mass index = 15) presented with dysphagia, coughing, and fever. Blood analysis showed elevated inflammatory parameters (C-reactive protein = 32.5 mg/dL, 30 700 leukocytes/μL), accompanied by rapid clinical deterioration. Computed tomography (CT) (Fig 1) suggested an esophageal rupture with pneumomediastinum, right-sided pleural effusion, and intrapulmonary abscess formation. Esophageal perforation resulting from advanced esophageal carcinoma was confirmed at emergency esophagogastroduodenoscopy and immediate surgical intervention was planned, as endoscopic treatment was not appropriate because of progressive septic aggravation. The pleural cavity was explored via a right-sided anterolateral thoracotomy. At subsequent pleural adhesiolysis and debridement, the visceral pleura of the lower pulmonary lobe ruptured and the pulmonary abscess containing pus and solid pieces of food was opened and evacuated. Because the lower pulmonary lobe was transformed into a cavernous abscess formation, we decided to perform a complete resection of the lower pulmonary lobe. Subsequently, simultaneous esophagectomy was performed, resulting in cervical diversion and blind closure at the esophageal hiatus of the diaphragm. The postoperative course in the intensive care unit was characterized by

Keywords
Endoscopic stent placement; esophagopulmonary fistula; esophageal cancer; pulmonary abscess formation; surgical treatment.

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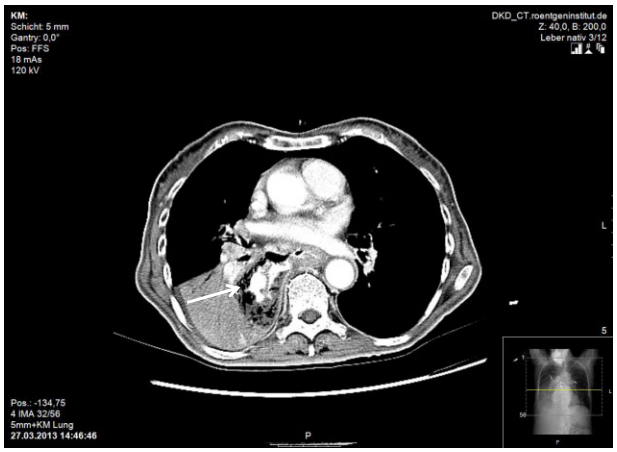

Figure 1 Computed tomography with oral contrast medium (mediastinal window) of the chest of case 1, showing an esophagopulmonary fistula with extravasation of oral contrast medium (arrow) with a lung abscess in the right lower lobe.

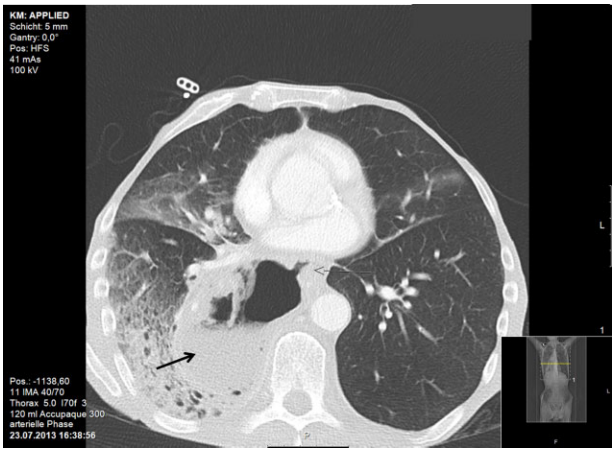

Figure 2 Computed tomography of the chest of case 2, showing an esophagopulmonary fistula (arrow) with a lung abscess in the right lower lobe (black arrow).

persistent pulmonary insufficiency requiring invasive mechanical ventilation.

Twelve days postoperatively an insufficiency of the bronchial stump required surgical revision and resection of the middle pulmonary lobe. The resulting staple line of the intermediate bronchus was buttressed with a pedicled intercostal muscle flap. Unfortunately the intensive care unit stay was complicated by recurrent infections in the remaining upper pulmonal lobe, which were caused by multiresistant Pseudomonas aeruginosa. This ultimately necessitated a pneumonectomy of the residual right lung. The remaining stump of the main bronchus was reinforced by a pedicled latissimus dorsi flap.

Three weeks after the pneumonectomy, the gastrointestinal passage was reconstructed using a retrosternal isoperistaltic colonic interposition and the residual tumor, which was detected in the histopathological investigation of the original esophageal specimen, was removed by performing a gastrectomy. This achieved a final staging of pT4b, pN1, L1, Pn0, G2, R0 (UICC IIIc).

Because of persistent weaning difficulties and respirator dependency, the patient was transferred to a specialized weaning clinic 15 weeks after the initial surgery.

A 60-year-old man presented with progressive dysphagia, coughing after fluid intake, and significant weight loss to the Gastroenterological Department of the University Hospital Düsseldorf. A histologically confirmed squamous cell carcinoma of the esophagus had been previously diagnosed. At admission, the patient had a body mass index of less than 15. Endoscopic examination confirmed a tumorous lesion in the distal esophagus with subtotal luminal obstruction and a mediastinal fistula. CT displayed a large abscess formation (Fig 2) nearly consuming the right lower pulmonary lobe, as well as an inflammatory involvement of the middle lobe. In the absence of septic aggravation, the patient underwent endoscopic placement of a covered self-expanding esophageal stent on the following day. Interventional CT-guided drainage of the pulmonary abscess formation was subsequently planned.

Two days after endoscopic stent placement, the patient developed an abdominal tenderness and signs of peritonitis. An emergent surgical intervention was performed and a perforated gastric ulcer was excised. Postoperatively, the patient was monitored in the intensive care unit and required increasing catecholamine support and re-intubation as a result of incipient pulmonary sepsis. With respect to the persisting pulmonary abscess formation and the underlying tumorous lesion of the distal esophagus without distant metastasis, the option of definitive surgical treatment consisting of esophageal and simultaneous pulmonary resection was suggested, and the patient and his family gave informed consent. Subsequently, elective resections of the lower pulmonary lobe and the thoracic esophagus were performed simultaneously with cervical diversion and distal closure. The middle pulmonary lobe could be preserved with clear margins at frozen-section analysis.

Postoperatively, the patient gradually improved, though he persistently depended upon mechanical ventilation. Three weeks later the gastrointestinal continuity was reconstructed using an isoperistaltic colonic interposition. The stomach, which was affected by several persisting ulcers, was removed. According to histopathological evaluation of all surgical specimens, the squamous esophageal carcinoma was finally staged pT4b, pN1, L0, Pn0, G2, R0 (Union for International Cancer Control IIIc).

The further postoperative course was prolonged and characterized by difficult weaning as a result of respiratory muscle

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weakness, as well as occasional episodes of aspiration requiring persistent use of the tracheostoma. As a result of intensive physiotherapeutic exercise the patient could finally be weaned from the ventilator during the daytime, enabling placement of a tracheotomy speaking valve. Continued ventilation was required at night to avoid respiratory muscle exhaustion.

Five weeks after reconstruction of the gastrointestinal continuity he was discharged and transferred to a rehabilitation center for weaning and physiotherapeutic mobilization.

Follow-up analysis revealed that both patients died at eight and seven months after the initial operation, respectively.

Discussion

The incidence of malignant esophagorespiratory fistulas in esophageal cancer patients is generally considered to be in the range of 5% to 13%.[9,10] Most of the lesions are localized between the esophagus and the tracheobronchial tree, but a subgroup (3–11%) presents with direct fistulas into the pulmonary parenchyma forming an inflamed necrotic alveolar cavity, resulting in extended pulmonary abscess formation.[2,9,11] Treatment of this devastating condition is highly individual. Therapeutic modalities comprise esophagopulmonary resection, endoscopic placement of self-expandable covered stents, esophageal diversion, and non-invasive supportive therapy. Most patients primarily undergo endoscopic stent placement, reflected in the numerous publications available. Surprisingly, in most reports, the option of surgical resection enabling the removal of both septic necrosis and malignant tissue is not even mentioned, although the tumor is frequently localized without signs of distant metastasis.[12]

Unlike esophagotracheal fistula, the paramount issue of esophagopulmonary fistula is the life-threatening necrotizing pneumonitis with large pulmonary abscess formation. Surgical therapy and resection of all gangrenous lung parenchyma should be the treatment goal if therapy is desired and the patient's general condition allows it.[13,14] Stent placement might even be detrimental in the presence of pulmonary abscess formation, because the natural drainage of the resulting cavern into the esophagus would be prevented by the stent placement. In a recent report of 14 patients who underwent palliative treatment with endoscopic stent placement, all died within a mean observation period of 65 days.[3]

The results of surgical resection in the two cases presented compare favorably with these results. Even though both patients had considerably reduced general health and pre-existing pulmonary co-morbidity, they survived various surgical interventions and were discharged tumor free and with restored swallowing function, 92 and 114 days after primary surgery, respectively.

Esophagopulmonary fistula is a dreadful complication of the underlying malignancy and the two cases presented impressively demonstrate the difficulties associated with major surgery when performed in such patients. However, the therapeutic option of surgical resection should at least be considered, particularly in patients who are free of distant metastasis (M0).

However, if palliation is the predominant goal of treatment, major surgery could be of limited benefit because a prolonged hospital stay and surgery-related morbidity must be assumed. On the other hand, in the presence of pulmonary abscess formation, the achievements of non-surgical management might also be limited because the fundamental principle "*ubi pus ibi evacua*" is disregarded, and patients rapidly deteriorate as a result of pulmonary sepsis.

Disclosure

No authors report any conflict of interest.

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